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Case Report

Surgical management of a large palatal pleomorphic adenoma by intra oral route: a difficult case

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ABSTRACT

Despite being the most common benign tumor of salivary glands, pleomorphic adenoma in hard palate is a rare site. Such tumors are a challenge to remove surgically. These tumors usually require an extensive resection with wide margins and removal of surrounding mucoperiosteum. There is also usually requirement of removal of a number of teeth or portion of maxilla, especially if the size is huge. The resulting disfigurement can be a problem for young females. We present a case of successful treatment of pleomorphic adenoma of the hard palate in a 15-year old female patient, which was achieved through an enbloc removal technique with clear margins, without the need for partial maxillectomy or bone removal. This approach played a pivotal role in preventing any potential disfigurement and preserving the aesthetic appearance of the patient. The personalized approach towards patient care was instrumental in tailoring the surgical strategy to meet the specific needs of the individual.

Keywords: Palatal pleomorphic adenoma, Intra oral excision, Minor salivary gland tumor, Enbloc removal, Primary closure, Intra oral tumor

INTRODUCTION

Salivary gland tumors are uncommon and make about 2-3% of head as well as the neck tumors. A common benign tumor in parotid gland and other major salivary glands is pleomorphic adenoma.[1] However, the pleomorphic adenoma of hard palate minor salivary glands is a rather uncommon. On the hard palate, it typically presents as a slow-growing submucosal tumor.[2] Females are slightly more likely to develop pleomorphic adenoma of hard palate and incidence peaks during the third and fifth decades of life.[3] PA is primarily treated through surgical excision. Although pleomorphic adenoma is a benign tumor, they often have microscopic pseudopod-like extensions into the surrounding tissue due to the "dehiscence" in the capsule. Hence, the resection of the tumor with an adequate margin of grossly normal surrounding tissue is required to the prevent local recurrence despite the fact that they are usually well-encapsulated.[4]

CASE REPORT

A 15-year-old female presented to the department of ENT with complains of a painless swelling on hard palate, which was slow-growing. Intraoral examination revealed a painless, immobile, firm and nodular lesion, forming a bulge. The size of this lesion was about 5 cm in diameter. The tumor was covered by healthy, smooth and intact mucosa which was normal in color. No ulceration of overlying skin was noted. The patient did not complain of any pain or tenderness although patient had difficulty in speaking and swallowing.

Examination of neck nodes revealed no nodal enlargement. FNAC of the swelling was advised.

Cytology smears were cellular and showed epithelial, myoepithelial and stromal components. The epithelial components formed clusters and few acini. The myoepithelial cells were mostly spindloid and were seen merging with fibrillary myxoid stroma.

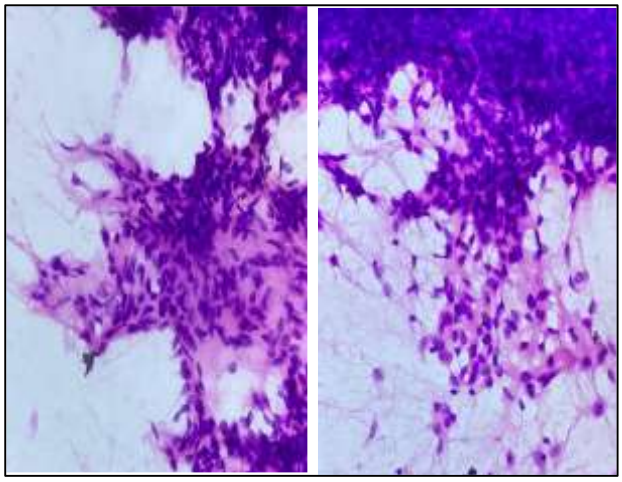

Figure 1: FNAC smears on H/E stain show epithelial and myoepithelial compinents. Epithelial component forms vague acini and myoepithelial cells appear bipolar/spindloid. Stromal fragments are also noted containing many myoepithelial cells.

Contrast CT scan of face showed expansile lesion measuring about 5×4 cm, noted in posterior aspect of left maxillary sinus with involvement of soft palate. Intraoral extention of the lesion was noted. Rest of the visualized paranasal sinuses otherwise appeared normal. No bony erosions were identified. Possible diagnosis of pleomorphic adenoma of soft palate was given.

By carefully assessing the tumor characteristics, considering the patient's age, and analyzing the potential functional and cosmetic outcomes, the surgical team was able to devise a treatment plan that focused on achieving optimal results while minimizing any adverse effects.

Surgery

Excision of the mass was done under general anesthesia. An incision is made intraorally and Mucosa around the lesion was marked and incised using the surgical blade. Mucosal flaps were elevated. The aim was to provide optimal access to the tumor while minimizing trauma to adjacent tissues. The surgeon carefully dissected and separated the tumor from the surrounding tissues, taking care to maintain clear surgical margins of 0.5 to 1 cm. The whole encapsulated tumor mass was excised from all around along with circumferential removal of soft tissue. This step required meticulous technique and attention to detail to prevent any damage to vital structures and ensure complete removal of the tumor. Posterior part of the tumor was deep seated hence its removal was challenging. The underlying bone was expanded but careful examination revealed it was not eroded. The tumor was removed enbloc by careful removal of intact capsule Hemostasis was achieved and the wound was closed using sutures an intraoral pack was kept for exerting pressure over the wound. The patient was monitored post operatively and was stable for remaining stay at hospital. Pain management and appropriate postoperative instructions were provided. Follow-up appointments were scheduled to monitor healing and assess for any signs of recurrence.

Figure 2: Intraoral bulge of tumor.

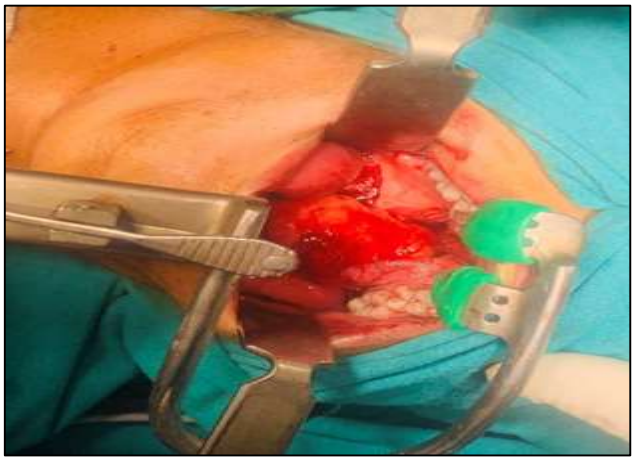

Figure 3: Tumor being carefully resected after raising the mucosal flaps.

Figure 4: Resected tumor.

The tumor was sent to pathology department. Gross examination showed an encapsulated nodule with

surgically intact surface, measuring 5.5×3.5×3.5 cm with solid whitish and glistening cut surface.

Microscopy showed a benign encapsulated tumor composed of cellular and stromal elements.

The capsule was extensively sampled to examine any surgical or anatomical breach. At all places, the capsule was found to be intact. However, the thickness of capsule was variable.

The cellular elements are composed of epithelial and myoepithelial cells. The epithelial cells form benign glandular acinic structures and few sheets. The myoepithelial cells are spindloid. Stroma is myxoid to chrondromyxoid. Myopeithelial cells are seen merging into the stromal components.

Focal areas showed formation of mushroom like projections of the tumor into the capsule. However, no separate satellite nodule was found.

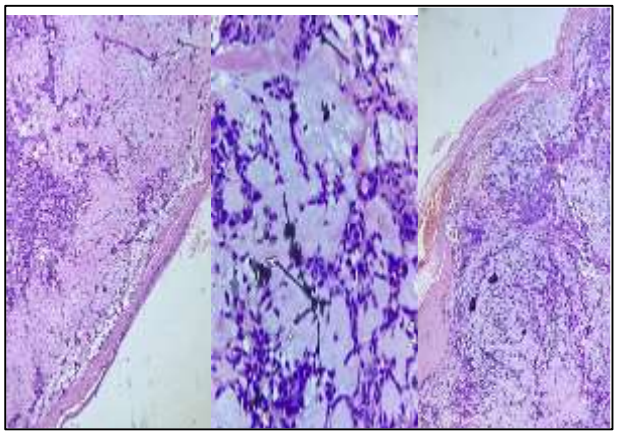

Figure 5: Microphotographs of histopathological examination revealed intact surgical capsule. The underlying tumor showed benign epithelial and myoepithelial components. Epithelial component formed acini and sheets. Myopepithelial component is formed by spindloid cells which are seen merging into the chondromyxoid stroma.

There is formation of a bulge in the tumor, however no breach of capsule is identified.

Post-operative follows up showed no evidence of recurrence.

This case highlights the significance of personalized care in the field of surgery, emphasizing the importance of considering individual patient factors and preferences to ensure the best possible outcomes.

DISCUSSION

According to the world health organization, pleomorphic adenoma is a tumor that is localized and has pleomorphic or mixed epithelial origin. Pleomorphic adenoma is one of the commonest tumors of salivary glands. Up to two-thirds of all salivary gland neoplasms are pleomorphic adenomas. Willis coined the term "pleomorphic adenoma". It was previously known by other names such as mixed tumor, enclavoma, branchioma, endothelioma, and endochroma etc.5 Even though pleomorphic adenomas are the commonest tumors of salivary glands, minor salivary glands of hard palate are not frequently involved.[2]

Clinically, patient usually presents with a slow growing, firm or rubbery submucosal mass without ulceration or surrounding inflammation on hard palate. Pain and tenderness are usually not seen.[2] For diagnosis of pleomorphic adenoma, history, physical examination, radiological evaluation and pathological evaluation, all play an important role.[6]

Computed tomography (CT) and magnetic resonance imaging (MRI) are commonly used to confirm the presence of the tumor.[6] The most accurate way to evaluate palatal tumor bone invasion and show how it spread to the maxillary sinus or nasal cavity is with computed tomography (CT).[7] MRI provides more precise information on the size and makeup of the tumor. MRI is also the most effective radiological diagnostic technique for finding recurring tumors as it makes the difference between a tumor and fibrosis more obvious.[7] Another additional benefit is that MRI does not expose patients to radiation.[7] However, histopathology is used to make the final diagnosis. However, pathological evaluation like fine-needle aspiration biopsy (FNAB) is needed to distinguish between malignant and benign lesions.[6]

Histologically pleomorphic adenoma is composed of a well-defined neoplasm with a fibrous connective tissue capsule and cells with ductal and myoepithelial. As a result of the presence of myxoid, ductiform, hyaline, plasmacytoid, osteoid, and chondroid regions, the phrase "benign mixed tumor" is also used to describe it.[3] A special attention should be paid to the specific characteristics of the capsule likely to be associated with possible recurrence, such as the presence of a complete and intact tumor capsule and surgically induced capsular defects and features like satellite nodules and pseudopodia.[8] An anatomically intact fibrous capsule that completely encases the tumor tissue is referred to as a full capsule. A pseudopodium is a secondary nodule that has been "re-encapsulated" and is localized within the primary tumor capsule, isolated from the tumor mass only by a layer of fibrous capsular tissue.[8] In our case, the capsule and margins were examined carefully. The capsule of our patient was surgically intact and complete capsule was found on microscopic examination. Presence of pseudopodia was not noted.

Treatment of choice for pleomorphic adenoma is surgical excision, especially in cases of PA of minor salivary glands. It is recommended to establish a margin of 5 mm

in small salivary glands. Enucleation is usually not regarded as the only option because of the risk of local recurrence.[9] To reduce recurrence and the likelihood that the lesion will turn malignant, the surgeon should attempt to prevent any breaks in the lesion's continuity and remove the entire lesion in one go.[6] In our case, the whole tumor was removed in Toto with clear margins and intact capsule. It is also recommended that to prevent recurrence, the underlying bone should be curetted with a sharp spoon or bur while being thoroughly irrigated with sterile normal saline.[2] In our case, there was no erosion of the hard palate, hence we did not do curettage of the bone.

Following a thorough surgical excision, recurrences are extremely rare. Recurrence, if it happens at all, may be caused by poor surgical procedures such leaving behind tiny pseudopod-like extensions, capsular rupture during surgery and tumor rupture with tumor cell spilling.[2] In our case, circumferential soft tissue margins were adequately removed with careful surgical preservation of capsule. Extensive sampling of margins and capsule revealed margins free of tumor, intact capsule and no pseudopodia. No recurrence was seen after 6 months of monitoring, although it is recommended that patient should be kept on long-term follow-up.

CONCLUSION

In conclusion, the successful surgery to remove the palatal pleomorphic adenoma in this young 15-year-old female patient demonstrated the effectiveness of complete enbloc resection with clear margins. The meticulous surgical technique employed during the procedure ensured the complete excision of the tumor with intact capsule and clear surgical margins minimized the risk of residual or recurrent disease. This surgical approach not only achieved the primary objective of tumor removal but also preserved the integrity of the maxilla and teeth, preventing any disfigurement. Preserving the maxilla and teeth played a vital role in maintaining the patient's oral function and aesthetics, which was a major concern for the patient and her family. This approach not only addressed the medical aspect of the case but also considered the patient's quality of life and psychological well-being.

The absence of recurrence during the six-month follow-up period reinforces the positive prognosis, bringing hope and optimism to both the patient and the healthcare team.

However continuous surveillance and regular follow up visits have been advised to monitor the patient's long-term progress and ensure the absence of any potential recurrence or complications.

This case serves as a testament that the advancements in surgical techniques and personalized treatment strategies can lead to successful outcomes in the management of palatal pleomorphic adenoma.

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REFERENCES

1. Rahnama M, Orzędała-Koszel U, Czupkałło L, Lobacz M. Pleomorphic adenoma of the palate: a case report and review of the literature. Contemp Oncol (Pozn). 2013;17(1):103-6.
2. Patigaroo SA, Patigaroo FA, Ashraf J, Mehfooz N, Shakeel M, Khan NA et al. Pleomorphic adenoma of hard palate: an experience. J Maxillofac Oral Surg. 2014;13(1):36-41.
3. De Lima, Fidêncio F, Pires BC; Rocha, Caroli A, Solani MI. Surgical Management of Palatal Pleomorphic Adenoma (PPA) Recurrence After 10 years, Treated at a Brazilian Center-A Case Report. Ann Maxillofacial Surg. 2022;10(2):533-6.
4. Arumugam P, Christopher P J, Kumar S. Pleomorphic Adenoma of the Palate: A Case Report. Cureus 2019;11(3):e4308.
5. Almeslet AS. Pleomorphic Adenoma: A Systematic Review. Int J Clin Pediatr Dent. 2020;13(3):284-7.
6. Yousra Z, Saliha C. Pleomorphic adenoma of hard palate: a case report. Pan Afr Med J. 2021;38:146.
7. Acaban MB. Importance of Differential Diagnosis of Palate Pleomorphic Adenoma; Case Report and Literature Review. Res Square. 2023;1.
8. Mantsopoulos K, Sievert M, Iro A-K. Histopathological comparison of pleomorphic adenomas of the parotid and submandibular gland. Oral Dis. 2022;28:1131-6.
9. Ordóñez JS, Arévalo MA, Torres MF. Pleomorphic adenoma of minor salivary glands: Review of the literature. World J Advanced Res Rev. 2023;18(02):365-74.

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